A systematic review of vestibular stimulation in cerebral palsy

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Abstract

Purpose

Identify the types and dosage of vestibular stimulation interventions in persons with cerebral palsy (CP), and establish the efficacy of these interventions on balance and function.

Materials and Methods

This systematic review followed Preferred Reporting Items for Systematic Review and Meta-Analysis Protocols to search for studies evaluating vestibular stimulation interventions in persons with CP. Information sources included MEDLINE, Embase, CINAHL, Cochrane Central Register of Controlled Trials, clinicaltrials.gov and the World Health Organisation registry. Methodological quality was assessed by two independent reviewers using the Methodological Index of Non-Randomised Studies (MINORS) and Cochrane Risk of Bias Tool.

Results

Five articles were included. Three randomised studies were judged to have high risk of bias in at least one domain of the Cochrane Risk of Bias Tool. Two non-randomised studies were rated as low methodological quality using the MINORS tool. All studies used exercise-based vestibular stimulation, but there was little homogeneity regarding dosage. Findings related to efficacy of vestibular stimulation were inconsistent.

Conclusions

Clinical practice recommendations cannot be made due to lack of high quality studies and heterogeneity of treatment protocols. Future research should address theory-driven selection of intervention, establish dosage, use psychometrically robust tools and include all ages of persons with CP.

Keywords

Vestibular stimulation; cerebral palsy; balance; rehabilitation; function

Word count

3541

Implications for Rehabilitation

- Optimal intervention parameters for vestibular stimulation cannot be determined from existing literature.
- Further studies to describe vestibular stimulation intervention components and duration are warranted.
- In practice, use of valid and reliable balance and gross motor function outcome measures are essential if using vestibular stimulation techniques with people with CP, as the efficacy of these interventions has not been clearly demonstrated.
- Investigation of electrical Vestibular Nerve Stimulation in people with CP is warranted.

Introduction

Cerebral palsy (CP) is a permanent disorder of posture and movement caused by disturbances in the developing brain [1]. It is the most common form of childhood physical disability, affecting approximately 1 in every 500 children internationally [2-4]. Whilst approximately 60% of children with CP can walk independently[5], many experience decreased postural stability [6-7], defined as the ability to control the centre of mass relative to the base of support [8].

Decreased postural stability in persons with CP affects gross motor skills such as walking, running and jumping [9-10], leading to difficulties with activities of daily living, participation in sports and leisure activities, quality of life and social interactions [6,10-11]. In addition, impaired postural stability increases risk of falling during walking [12-13] which in turn carries associated physical and psychosocial consequences [14]. Therefore, interventions targeting postural instability and balance deficits warrant investigation in people with CP.

Given that the vestibular system transmits sensory information to the brain via the vestibular nerve to maintain postural stability, stimulation of this system may reduce balance deficits in children and adults with CP. Indeed, vestibular stimulation via specific exercises such as spinning and swinging has demonstrated improvements in postural stability, specifically, static and dynamic balance [15], and in sitting balance [16] in children with CP. Possible mechanisms of action include maturation of the vestibuloocular reflex, thus enabling stable retinal image during head movements [17] and impacts on the lateral vestibulospinal tract, facilitating maintenance of upright and balanced posture [18].

Alternatively, the vestibular system can be stimulated by way of electrical current delivered via self-adhesive pads on the mastoid processes, frequently known as Galvanic Nerve Stimulation [19-24] or Vestibular Nerve Stimulation (VeNS) [25]. Early clinical research has reported that VeNS improves postural stability in persons with Parkinson's Disease [19-21], bilateral vestibulopathy [22], and the elderly population [23-24], potentially via vestibular neuroplasticity and enhanced vestibular information processing [23], however there is a paucity of evidence in people with CP.

Although various methods of vestibular stimulation are available, information relating to the dosage and efficacy of each is not well documented. Therefore, this systematic review aimed to (i) identify the types and dosage of vestibular stimulation interventions used in the treatment of balance and associated postural and functional deficits in persons with CP, and (ii) establish the efficacy of vestibular stimulation interventions on balance and function in this population.

Materials and Methods

Study design

A systematic search and narrative literature review were undertaken, compatible with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses Protocols (PRISMA-P) [26]. The review was registered with PROSPERO (Reference: CRD42019140462).

Search Strategy

The following electronic databases were searched from database inception to 21st January 2020: Medical Literature Analysis and Retrieval System Online (MEDLINE), Embase and Cumulative Index to Nursing and Allied Health Literature (CINAHL). Trial registries including Cochrane Central Register of Controlled Trials, clinicaltrials.gov and the World Health Organisation (WHO) registry were also searched. Hand searching of the reference lists of relevant systematic reviews returned by the primary search was also undertaken.

Search terms

The PICO (Patient, Intervention, Control/Comparison, and Outcome) model was used to tailor a search strategy with individual search terms [27]. Due to the paucity of literature in the area, a simple search strategy was adopted: only the terms 'cerebral palsy' AND 'vestibular' were searched as keywords.

Eligibility criteria

Inclusion and exclusion criteria were based on study design, participant diagnosis and the outcomes of interest to the review. All quantitative study designs were included; systematic reviews were excluded but reference lists were searched to ensure inclusion of all relevant primary research. All study participants had a clinical diagnosis of CP and no limits were set on age, gender, type or severity of the condition. All types of vestibular stimulation interventions were eligible for inclusion in this review. In line with the aims of the review, the primary outcome of interest was the type of vestibular stimulation, including administration and duration (e.g. time administered and length of intervention period). Secondary review outcomes were the effects of vestibular stimulation on posture, balance and function. Articles published in languages other than English were excluded due to lack of translation facilities.

Screening and selection

One reviewer (DT) completed all electronic searches. Titles and abstracts identified by the initial search strategy were screened by two independent reviewers (DT and KMC) to determine eligibility. When the title and abstract did not clearly indicate whether or not a study should be included, the full text was obtained and assessed for eligibility by two members of the review team (DT and KMC). Where disagreements occurred, consensus was reached by discussion with a third reviewer (CK).

Data extraction and management

Results from searches and all retrieved references were imported and managed in RefWorks reference management software (available at: https://refworks.proquest.com). A data extraction form was developed a priori by the research team. The form was piloted and modified in advance of formal data extraction commencing. Data from eligible studies were extracted independently by two reviewers (DT and KMC).

Data extracted included descriptive information about the study (e.g. design, sample size, and setting), demographic information on the participants (e.g. gender, age) and description of CP (e.g. type and severity). Further data extracted included a description of the intervention (and control), the outcome measures employed, study results, adherence to the intervention and adverse events.

Risk of bias

Each included study was independently assessed for risk of bias by two reviewers (DT and KMC). The Methodological Index of Non-Randomised Studies (MINORS) [28] was used to describe methodological quality of non-randomised studies. The MINORS consists of 12 domains, each scored as 0 (not reported), 1 (reported but inadequate) or 2 (reported and adequate), giving a total score of 24, with a higher score indicative of a lower risk of bias in the study. In non-comparative, non-randomised studies, four domains of the MINORS tool are not scored, resulting in a maximum possible score of 16 for these studies.

Methodological quality of randomised studies was assessed using the Cochrane Risk of Bias Tool 2.0 [29-30]. This tool is structured into five mandatory domains of bias focusing on different aspects of trial design, conduct and reporting. An algorithm based on responses to the questions within each domain is provided to determine the domain of interest as ‘low’, ‘high’ or ‘some concerns’ relating to risk of bias. Inconsistencies were resolved by discussion and inclusion of a third reviewer (CK).

Results

Search strategy results

Electronic database searches returned 130 potentially eligible studies (figure 1). A further two studies were located by hand searches of reference lists of eligible studies from the initial search. After removing duplicates (n=66), 66 studies remained. Following screening by title and abstract, 14 studies required full text review to determine eligibility for inclusion. Of these, only five met all inclusion criteria and were retained for review [15-16, 18, 31-32].

[Figure 1 near here]

Summary of included papers

Of the five included papers, three were randomised controlled trials (RCTs) and two were non-randomised studies. All evaluated the efficacy of vestibular stimulation in children with CP. One of the three randomised studies used a matched pairs design to establish effect of vestibular stimulation on motor behaviour in children aged between eight and 56 months [32]. The second randomised study employed a longitudinal, cross-over design evaluating the effect of vestibular intervention on gross motor function in children with an average age of 7.5 years [15]. The remaining randomised study was a double blind RCT evaluating the effect of vestibular stimulation on quantitative measures of postural stability (assessed using a force plate) in children aged 3-10 years [18]. Of the two non-randomised studies, one was a controlled trial evaluating the effect of vestibular stimulation on gross motor skills in pre-ambulatory children aged 2-6 years [31], and one was a single case report on the effects of vestibular stimulation on a 19 month old child with hypotonic CP [16].

Notably, two of the studies were conducted around 40 years ago in the USA [31] and Australia [32]. In contrast, the three remaining studies were published in the last five years [15-16, 18] and were undertaken in Iran [18], Korea [16] and Italy [15]. Of the three included studies that provided participant details in relation to type of CP [15-16, 32], none were homogenous in motor type or distribution of CP. Ages of children across studies ranged from 19 months [16] to 10 years old [18]. Full details are available in table 1.

[Table 1 near here]

Types and dosage of vestibular stimulation

All of the included studies stimulated the vestibular system via swinging, rotation or spinning movements in different postures (see table 1). Three of the five studies investigated vestibular stimulation activities in isolation [16, 31-32]. Of these, two employed spinning movements in sitting and side-lying in a hand-operated rotating chair [31-32], whilst one used a swinging protocol that progressed from lying on an infant swing to standing on a platform swing [16]. In contrast, two studies incorporated vestibular stimulation activities into conventional therapy sessions [15, 18]. For example, during the last 20 minutes of a 45 minute occupational therapy session, Hosseini et al (2015) [18] used different equipment (e.g. tilt boards, scooter boards, CP balls and spinners) to achieve four types of vestibular stimulation: anteroposterior tilts, lateral tilts, ascending-descending orientation with gravity, and spinning. Similarly, Tramontano et al [15] added three types of vestibular exercises into a tailored neurodevelopmental therapy session. The vestibular stimulation exercises included gaze stability training in a darkened room, gait training, and seated rotation activities on a backless stool [15].

There was little homogeneity regarding dosage of vestibular stimulation activities. Similarly, each study had a unique intervention protocol that was described in varying detail in the included papers. Intervention frequency and duration varied from 10 sessions over five weeks [15], 12 sessions over six weeks [18], 16 sessions over four weeks [31-32], and 30 sessions over 10 weeks [16]. Individual session duration was difficult to establish in most cases due to insufficient reporting, but was estimated to range from approximately 10 minutes (10 x1 minute spins) [31-32] to one hour [16] per session.

Efficacy of vestibular stimulation interventions

Efficacy of vestibular stimulation was evaluated using a number of different outcome measures as detailed in table 1. These included functional tests such as the Motor Skills Test [33] (adapted by Chee et al 1978 [31]), Reflex Test [34] used by Chee et al (1978 [31]), Bayley Scales of Infant Development [35-36] used by Sellick & Over (1980 [32]) and An (2015 [16]), and the Gross Motor Function Measure (GMFM [37]) used by Tramontano et al (2017 [15]). A patient-centred measure, the Goal Attainment Scale [38], was used by one of the five included studies [15]. In addition, two of the included studies employed displacement measurements, carried out with a triaxial accelerometer [15] and force plate [18], as quantitative indicators of postural control.

Results from the included studies are summarised in table 1. A matched pairs RCT [32] with 10 participants in each group reported no significant improvement on the Bayley Scales of Infant Development as a result of vestibular stimulation. Similarly, in another randomised study, Hosseini et al (2015 [18]) demonstrated no significant difference between treatment (n=8 participants) and control groups (n=8 participants) post-intervention in quantitative measurements of postural stability, with the exception of a significant improvement in the ability to change and control centre of pressure displacement faster (p=0.036). The remaining randomised study [15], a controlled crossover trial including 14 participants, reported varying within group differences across outcome measures. A significant improvement was noted in the GAS-score following neurodevelopmental therapy combined with vestibular stimulation (p=0.003), but both neurodevelopmental therapy combined with vestibular stimulation and neurodevelopmental therapy alone demonstrated significant improvement in GMFM scores (intervention group p=0.005, control group p=0.034). Both non-randomised studies reported positive effects as a result of their vestibular stimulation interventions. Chee et al (1978 [31]) detected a significant difference between intervention (n=12 participants) and control (n=11 participants) groups in the Motor Skills Test (p<0.01) and Reflex Test (p<0.001); and An (2015 [16]) reported improvements in motor and mental scores on the Bayley Scales of Infant Development (raw scores increased from 40 to 58, and 21 to 46, respectively) in one child with hypotonic CP.

Risk of bias assessment

As shown in table 2, the overall scores for all three RCTs included in this review indicated a high risk of bias in at least one domain on the Cochrane Risk of Bias Tool [15,18, 32]. There were some concerns raised regarding domain 1 (risk of bias arising from the randomisation process) in two of the three randomised studies [18, 32]. In all three studies, there were some concerns of risk of bias from effect of assignment to intervention (domain 2a), due in part to the difficulty of blinding such treatment allocation. In addition, high risk of bias was evident for domain 2b (effect of adhering to intervention) in all of the included randomised studies. There were also some concerns of risk of bias regarding selection of the reported result (domain 5). In contrast all three randomised studies were judged as having low risk of bias due to missing outcome data or in measurement of the outcome (domains 3 and 4).

[Table 2 near here]

Of the non-randomised studies included in the review, Chee et al (1978 [31]) scored 11/24 and An (2015 [16]) scored 8/16 on the MINORS scale (table 3). Both non-randomised studies had a clearly stated aim, endpoints were appropriate to the study aim and the follow-up period was appropriate. In contrast, four MINORS items were not reported by either study: these related to inclusion of consecutive patients, prospective collection of data, unbiased assessment of the study endpoint, and prospective calculation of study size. The four additional MINORS criteria employed for comparative studies were partially met by Chee et al (1978 [31]), however they failed to report an adequate control group and baseline equivalence of groups. There was perfect agreement between the independent reviewers in relation to the appraisal of risk of bias in the included studies.

[Table 3 near here]

Discussion

This systematic review of the types and effects of vestibular stimulation on posture, balance and function in children and adults with CP identified just five eligible studies [15-16, 18, 31-32]. Two of these studies were published over 40 years ago [31, 32] and the remaining three within the past five years [15, 16, 18]. It could be that this renewed interest in vestibular stimulation is associated with technological advances; whilst this was not apparent in the method of vestibular stimulation employed by recent studies, it was evidenced in the selection of technology dependent quantitative measurement tools such as force plates [18] and accelerometers [15]. All studies were conducted in children with CP and used exercises and movements, such as spinning and swinging, in an effort to stimulate the vestibular system and impact on postural stability and motor function. Study findings were not unanimous in their conclusions and methodological concerns regarding the conduct of included studies were identified, suggesting that there is a high risk of bias in the included studies. This means that it is not currently possible to endorse or refute the use of vestibular stimulation to improve balance, posture or function in people with CP.

This review identified the lack of a 'standardised' vestibular stimulation intervention, in terms of stimulation type (beyond spinning and swinging exercises), and duration of the intervention. Included studies did not clearly define vestibular stimulation, and did not provide a theory-driven approach to justify individual intervention choices. For example, clear reasoning for the type of stimulation, number of sessions and total duration of treatment was lacking, other than longer interventions aligned with the child's conventional therapy session schedule. Furthermore, only two of the included studies [31-32] appeared to use comparable vestibular stimulation interventions based on those originally described by Clarke et al (1977 [17]). Consequently, optimal intervention parameters for vestibular stimulation cannot be determined from the studies included in this review, and further studies that clearly describe the intervention components and duration are warranted. In addition, it was noted that none of the eligible studies investigated vestibular stimulation by delivery of electrical currents to the vestibular nerve via electrodes placed on the mastoid processes (Vestibular Nerve Stimulation, VeNS). Whilst VeNS has not yet been evaluated in people with CP, early clinical research in patients with Parkinson's Disease [19-21], bilateral vestibulopathy [22], and the elderly population [23-24] have reported significant improvements in postural stability. In addition to potential improvements in postural stability in people with CP, VeNS may be better tolerated than traditional spinning exercises and thus may provide a non-invasive alternative or adjunct to conventional therapies that can be delivered in the home setting. Research is warranted to evaluate the acceptability and feasibility of using VeNS as a treatment for balance deficits in persons with CP.

Overall, studies relating to the efficacy of vestibular stimulation reported conflicting results and conclusions. Whilst a high risk of bias was identified in all of the included randomised studies, they were considered to be of higher methodological quality than the non-randomised studies due to use of a more robust study design. Interestingly, the randomised studies reported no [32], or minimal, significant improvements [15, 18] following vestibular stimulation, whilst the non-randomised studies reported positive effects of the intervention [16, 31]. Therefore, study design and quality may have affected study findings. Poor methodological quality coupled with the paucity of studies to support the effectiveness of vestibular stimulation suggests that an insufficient body of evidence currently exists to make clinical practice recommendations.

In addition, variation in outcome measures employed by the included studies meant that synthesis of results was not possible, despite detection of significant improvements [31] on some standardised functional outcome measurement tools (the Motor Skills Test [33] and Reflex Test [34]). Interestingly, a positive effect on Goal Attainment Scale scores [38] was detected, suggesting that vestibular stimulation may be a useful adjunct to improve patient-centred outcomes that are clinically meaningful to patients and families. Studies employing quantitative, laboratory-based measurements of balance [15, 18] also demonstrated statistically significant improvements following vestibular stimulation. This may be partially explained by their focus on specific components of balance as opposed to overall function, however, the clinical significance of these results has not been determined. Furthermore, clinical utility of such quantitative outcome measurement tools needs to be considered due to the requirement of specialised equipment and training.

Finally, this review aimed to identify the types and dosage of vestibular stimulation interventions, and efficacy of the same, in children and adults with CP. In particular, given that previous research suggests that electrical vestibular stimulation may improve balance in adults with other neurological conditions [19-21], similar clinical benefits may be found in adults with CP. However, none of the studies included in this review involved adults with CP. Consistent with this finding, the paucity of literature evaluating interventions to improve dynamic balance and walking in adults with CP has been previously reported [39-40]. This paucity of evidence coupled with the reported decline in balance and walking abilities in adults with CP in their twenties and thirties [14], suggests future research evaluating efficacy of balance interventions, such as vestibular stimulation, should include adults with CP.

Strengths and limitations

Strengths of this review include the use of a systematic search strategy including use of search terms as keywords to widen the scope of the review. No restrictions on date or location of the research were included in the search strategy. The review also considered all types of vestibular stimulation interventions. In addition, data extraction and quality appraisal of included studies were carried out by two independent reviewers between whom there was a high level of agreement in data extraction and quality scores. In spite of these strengths, this review is limited to studies published in the English language and the potential effects of vestibular stimulation related to domains other than balance, posture and function were not considered.

Conclusions

A small number of studies were identified that evaluated the efficacy of vestibular stimulation in people with CP, all of which included children only. Although the types of vestibular stimulation employed were limited to exercise and movement, with no studies evaluating electrical stimulation modalities, optimal treatment parameters could not be established due to the heterogeneity of intervention protocols employed. The contradictory results and conclusions reported by the included studies, coupled with their poor methodological rigour, mean that this review is currently unable to support or refute the use of vestibular stimulation to improve posture, balance or function of people with CP. Accordingly, recommendations for further research are broad but should address evidence-based and/or theory driven selection of a clearly described vestibular intervention (exercise or electrical stimulation), establish parameters relating to intensity, frequency and duration of vestibular stimulation, use psychometrically robust outcome measures, and include people with CP of all ages.

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Declaration of interest statement

Interest in this area resulted from conversations with Neurovalens, a company that have developed a headset to deliver electrical vestibular stimulation. Neurovalens were not involved in the design, conduct or reporting of this systematic review.

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Table 1: Summary of included articles

Author	Stimulation type	Stated Study Aim	Study design	Sample size (n=)	Age range	Dosage	Outcome Measures	Results
Chee et al 1978[28]	Spinning exercises on a chair	To establish whether semi-circular canal stimulation significantly improves gross motor skills in pre-ambulatory children with CP	Non-randomised Controlled Trial	Intervention n=12, control n=11 (handled control n=6, non-handled control n=5)	2-6 years	Session duration NR, 16 sessions over 4 weeks	The Motor Skills Test, Reflex Test	Significant difference between intervention and overall control groups in post treatment scores on the Motor Skills Test ($p<0.01$) and Reflex Test ($p<0.001$). No significant difference between two control subgroups.
Kenneth & Over 1980[29]	Seated rotation	To establish whether vestibular stimulation improves the motor behaviour of children with CP	Matched pairs RCT	Intervention n=10, control n=10	8-56 months	2 sessions per day separated by 30 minutes. 2 days per week, for 4 weeks	Bayley Infant Development Scales, 1st edition	No statistically significant improvement in motor competence.
Hosseini et al 2015[30]	anteroposterior, lateral, ascending-descending movements and spinning	To investigate the effect of vestibular stimulation, stimulating and integrating both vestibular and proprioceptive systems on centre-of-pressure parameters in children with CP	Double-blind RCT	n=20 initially, reduced to n=16 for inclusion. Intervention n=8, control n=8	3-10 years	45 minute sessions 2 times weekly for 6 weeks	Quantitative measurements using a force plate and calculated by Matlab software: Range for After (RFA) – range of anterior posterior displacement in Y axis; Range Side Way (RSW) – displacement in the X axis; Mean Velocity (MV) – division of displacement on numbers; area of centre of pressure – mean rate of COP	Significant improvement for RSW (eyes open, $p<0.03$), Area ($p<0.04$), RFA ($p<0.001$) and RSW (eyes closed, $p<0.002$). No significant difference between eyes open and closed, except in velocity parameter in intervention group ($p<0.05$).

							displacement in both X and Y axes	
An 2015[16]	Infant swing (4x swing variants for the child)	To present the effects of vestibular stimulation through the use of swings, on a child with hypotonic CP	Single Case report	n=1	19 months	1-hour session, 3 times weekly for 10 weeks	Bayley Infant Development Scales, 2nd edition	Raw mental score increased from score of 21 to 46 (increase of 3 months in developmental age). Motor raw score increased from 40 to 58 (increase of 4 months in developmental age).
Tramontano et al 2017[15]	Vestibular physical therapy	To assess efficacy of vestibular physical therapy, specifically designed for children with CP to obtain motor function improvement	Longitudinal, Randomised, Controlled, Cross-over Trial	n=14	NR	50 minute session, 2 times weekly for 5 weeks	GMFM, GAS, quantitative tests carried out with a tri-axial accelerometer	Within group differences: GMFM – significant improvement after both NDT (p=0.034) and NDT + VS (p=0.005), GAS – significant improvement after NDT + VS (p=0.003), Accelerations – significant interaction between therapy and body axis during forward movement (p=0.044).

Legend: n, number of participants; CP, cerebral palsy; NR, not reported; RCT, Randomised Controlled Trial; RFA, Range for After; RSW, Range Side Way; MV, mean velocity; COP, centre of pressure; GMFM, Gross Motor Function Measure; GAS, Goal Attainment Scale; NDT, neurodevelopmental therapy; VS, vestibular stimulation

Table 2: Risk of bias assessment for randomised studies (Cochrane Risk of Bias Tool)[25-26]

Author and year	Domain 1	Domain 2a	Domain 2b	Domain 3	Domain 4	Domain 5	Overall
Sellick & Over 1980[29]	Some concerns	Some concerns	High ROB	Low ROB	Low ROB	Some concerns	High ROB in at least one domain
Hosseini et al 2015[30]	Some concerns	Some concerns	High ROB	Low ROB	Low ROB	Some concerns	High ROB in at least one domain
Tramontano et al 2017[15]	Low ROB	Some concerns	High ROB	Low ROB	Low ROB	Some concerns	High ROB in at least one domain

Legend: ROB, risk of bias

Table 3: Risk of bias for non-randomised studies (Methodological Index of Non-Randomised Studies)[24]

Author & year	Research design	MINORS questions												Total score
		1	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**	**10**	**11**	**12**	
Chee et al 1978[28]	Controlled trial	2	0	0	2	0	2	0	0	1	2	0	2	11/24
An 2015[16]	Case report	2	0	0	2	0	2	2	0	n/a	n/a	n/a	n/a	8/16

Figure 1:

PRISMA flowchart of included and excluded articles